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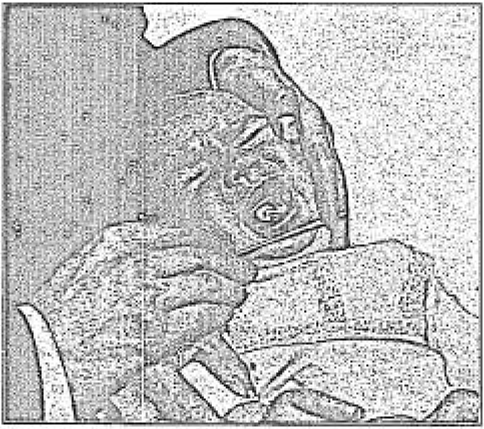
SUBLINGUAL GROWTH IN INFANTS.

(PRODUZIONE SOTTOLINGUALE, RIGA'S DISEASE, FEDE'S DISEASE, ETC.)

BY SAMUEL AMBERG, M.D.,
INSTRUCTOR IN PEDIATRICS, JOHNS HOPKINS UNIVERSITY.

IN December, 1902, a seven-months-old boy was brought to the Children's Department of the Johns Hopkins Hospital Dispensary on account of a tumor under the tongue. The parents of the baby are of American descent. The family history did not reveal any particular affections of the parents. There was particularly no suspicion of tuberculosis or syphilis. Four other children of the same parents never had anything like the affection which brought the baby to the dispensary. The general health was excellent and always had been, in spite of the fact that it enjoyed a rather liberal diet, after having been fed at the breast for about four months. There was no enlargement of the lymph glands of any region. The tumor underneath the tongue was about 1 to 1.5 cm. in diameter and about 5 mm. thick. It presented the appearance of a disk, was rather hard, and its surface was somewhat rough. The oral sufrace of the tumor was of a pearly-white



color, surrounded by a reddish margin; its centre was lightly depressed and showed some sloughing. The tumor was perfectly separated from the tongue and attached to the frenulum with a broad basis.

At the time the baby was seen the growth had persisted for about four weeks, having started, according to the mother's statement, as a hard, white pimple on the frenulum, at the time when the two median lower incisors made their appearance. These were the only teeth which had broken through. They pointed slightly backward, while their median side was slightly turned inward. Their cutting edge was somewhat ragged. Occasionally the tumor is said to have bled a little; it did so at the time of the second examination, after having been touched the day before with silver nitrate. It was possible to lift the growth a little, so that its removal could be readily accomplished by means of the scissors. The operation was performed about two months ago, and since that time nothing more was heard of the little patient.

The affection I have just described seems to occur most frequently in southern Italy, and the reports concerning it are mostly confined to the Italian literature. Since this case seems to be the first on record in the United States, it may be permissible to give a review of the literature. Several of the Italian authors mention an infection described by Urbino Cardarelli in 1857, under the name of "afta cachettica." Its chief characteristic seemed to have been an ulceration on the frenulum linguæ of infants, causing or inseparably associated with general wasting of the little patients, ultimately leading to the exitus lethalis. The notes given by Cardarelli are, according to Fede,[1] insufficient.

In 1872 Ridola[2] mentions a pathological condition of the mucosa under the tip of the tongue in babies of the first year of life, associated with protracted gastro-intestinal catarrh, anæmia, and weakness. The pathological change he describes as a necrosis resembling in its character a specific diphtheritic process. Its origin is attributed to compression brought about by the two median lower incisors. In 1875 Pandolfi[3] reported five cases of a special neoplasm under the tongue of suckling infants. It is described as a fibrous tumor reaching from the frenulum linguæ to the apex, round, of about 1 cm. diameter and 2 mm. thickness, hard, and of a pearly-white color. The rest of the mouth does not show any changes. General symptoms were associated with this tumor—nutrition was interfered with, the muscles were weak, the development retarded, and the mental faculties impaired. Two of the babies died. In one instance the tumor disappeared within a year and with it the general symptoms. In another case the disease, which had started in the sixth month, lasted until the fourth year. With the disappearance of the tumor the child regained its physical and mental health. Pandolfi thought to have observed a hitherto unknown disease. Independently of Pandolfi, Riga[4] laid down the outlines of a new malady in 1881, with the following characteristic features: Between the apex and frenulum linguæ or on other parts of the latter there is found what he called a neomembrane of a pearly color. It does not change its appearance and never occurs in other parts of the mouth. It is little or not elevated over the surrounding mucosa, of round shape, very difficult to detach, and if detached there is no (or very little) bleeding. After removing the neomembrane the surface on which it was implanted is bared of its mucosa. However strongly cauterized with silver nitrate, the neomembrane is reproduced after one or two days. It may grow to the size of about 1 cm. in diameter. There is no general or partial glossitis; the regional lymph glands are not enlarged. If suckling is interfered with, it is only on account of the weakness of the baby, whose general state of health is bad and becomes gradually worse. The end is nearly always death. In the first days the disease may perhaps be febrile, but later it is afebrile. Gastro-intestinal disturbances are nearly constantly present; rarely hemorrhages in the skin were observed. The disease occurs in suckling infants a little before or at the beginning of dentition. Riga is not inclined to regard the mechanical irritation produced by the teeth as an etiological factor. The parents are always well, but most of the babies present a status lymphaticus. He regards the disease as infectious, but not contagious, and the local lesion under the frenulum as of internal origin.

The communication of Riga was the starting point of a number of publications on the subject, and some of the authors described the affection under the name of Riga's disease. In the same year as Riga, Chiarello[5] reported several cases. In his conception the first manifestation is an ulceration, which later gives rise to diphtheritic products. The ulceration itself is brought about by mechanical irritation, caused by the movements of the tongue over the lower incisors in the act of suckling. If the ulceration begins before the eruption of the teeth, the sliding of the tongue over the gum, hardened through the underlying incisors, will cause an exfoliation of the epithelium and, subsequently, ulceration. Chiarello saw the disease only after the lower incisors had broken through, and always in the course of suckling. The season in which he saw it reached from summer to winter. The severe symptoms are regarded as a consequence of the ulceration. The disease is not infectious; the prognosis serious, although of his five cases one was cured completely; in one the growth had disappeared, but anæmia, etc., remained. The three other cases were still under observation, but did not show any alarming symptoms.

According to De Marinis,[6] the disease is not rare in his district, in southern Italy. The people there call it "ranocchia" (frog). He accepted this term, only translating it into Greek, "batracosi." It is described as a small, pearly-white, hard, painless fibroid neoformation under the frenulum linguæ. The neglected babies of the poor population are affected. The babies may apparently be in the best state of health or suffering from other diseases. The affection may cause general symptoms, finally leading to death. The most prominent symptom is a progressive cachexia due to mysterious reasons. Preceding, accompanying, or following the appearance of the batracosi are stomatitis, gingivitis, gastro-enteritis, etc. Anæmic and lymphatic conditions of the parents are regarded as predisposing and some significance is attributed to improper feeding; but external causes, as bad hygiene of the mouth, are more important etiologically than constitutional factors. The disease is a family disease in so far as members of one family are subjected to the same injuries. The prognosis is always serious.

Somma[7] sees in the lesion of the frenulum a pseudomembrane (mucositis fibrinosa frenuli linguæ), which starts as a dirty whitish spot, increases to a certain extent, and then remains stationary, with occasional exfoliations due to the action of the lower incisors. Five of his six patients were suffering from various serious diseases. He seems to think that the lesion of the frenulum is purely local and does not participate in the fatal result on its own account, but that this is due to an accompanying, more important disease, which need not be the same in every case. The babies are subjected to a more or less advanced cachexia and nearly constantly to gastro-intestinal disturbances. The disease is more frequent in summer time, when these latter are prevalent. Neither hereditary influences nor the infectious character of the lesion are regarded as proven. Whether or not the cause is mechanical is left undecided, although the lower incisors were present in all of the cases.

Jannone[8] reported an analysis of seventeen cases in 1884. The local lesion begins with an erosion of the inferior part of the tongue and the frenulum. Soon there develops a small, pearly-white, hard, painless spot, increasing to the size of a two-centesimo piece. It then becomes harder and thicker and can persist in this state for a long time. The tissue underneath the pseudomembrane consists of fibroid nodules. The pseudomembrane is difficult to detach and only in small pieces. When the membrane is removed the nodules bleed a little. He admits an individual and a family predisposition, relying on the history given by one of the mothers, who claimed to have lost three babies with the disease. The influence of the summer months is doubtless. In a cold climate the course of the disease is more rapid, the cure easier accomplished. The mechanical irritation produced by the incisors does not figure among the causes, but an influence of the dentition is recognized inasmuch as it affects the general health. The duration of the disease varies between eight days to twenty-one months, with alternating improvement and relapse. Any more serious manifestations are due to continuous disturbances of the digestion or to dyscrasic affections, which may occur during the disease. Only babies are affected who are in a precarious state of health; but the lesion of the frenulum, if it is not a direct consequence of another disease, is a complication of not much interest. Gastro-intestinal disturbances are constant companions of the disease; these are due to dentition, bad hygiene, and atmospheric conditions. When once cured Riga's disease may reappear. Of the seventeen cases two died, but their death is attributed to complications and negligence. The prognosis, therefore, seems to be favorable. Of interest are the experiments of the author. He put babies with Riga's disease to the breast of healthy women and healthy babies to the breast of women whose babies were affected with batracosi. Furthermore, he removed pieces of the pseudomembrane and placed them on the frenulum of healthy babies. The negative results of these experiments do not speak in favor of an infectious character of Riga's disease.

None of the authors thus far cited reported the histological examination of the growth nor results of an autopsy. The first who made a histological examination was Fede,[9] in 1890. He describes the lesion as a partial alteration of the mucous membrane of the mouth in the form of an excrescence under the frenulum linguæ of several millimetres' thickness and of about 1 cm. diameter. The surface is even or depressed from the margin toward the centre and sometimes it may be somewhat rough, like cauliflower. The tumor is rather hard and consistent, indolent, of a dirty, grayish-white color, with a pearly lustre. It may undergo necrobiosis and ulceration. Clinically Fede distinguishes three different forms. In the first the baby is perfectly healthy and the sublingual tumor is the only pathological manifestation present. In the second group the tumor is associated with other disturbances; the tumor does not improve as quickly and undergoes ulceration more easily. The third group includes those babies where in the course of a serious malady the baby acquires the "produzione sottolinguale." This is the form which is usually described as a grave disease, of which the tumor is said to be the local expression. But according to Fede, any disturbance which accompanies the sublingual growth is due to perfectly different causes and does not stand in any more intimate relation to the neoplasm. The only influence which he ascribes to these general disturbances he finds in the lessened resistance of the tissues of infants in a cachectic condition. The cause of the formation of the tumor is to be found in the mechanical irritation produced by the movements of the tongue of the suckling over the two median lower incisors, particularly when their edge is irregular, sharp, and ragged, taking into account a possible shortness of the frenulum.

If the tumor appears before the eruption of the incisors, the gum hardened by the underlying teeth takes their place, particularly when suckling is difficult. The course which the tumor follows can be different; improvement can alternate with retrogression until the tumor finally disappears completely. The histological examination of the first two tumors showed hypertrophy and hyperplasia of the epithelium and of the papillary body. Therefore, the neoplasm is called a papilloma. In later contributions Fede[10 11] reports the examination of fifteen of his own cases, one by Concetti, one by Giarrè, one by Nota, and two by Boccardi. The histological findings agree well. Fede describes a secondary change of the tumor, which he attributes to the persistency of the mechanical irritation. The epithelium undergoes necrosis and is then given off, while the still hypertrophic and hyperplastic papillary body is infiltrated with a grand mass of round cells, which are embedded in a network of fibrin. Thus the tumor gains the aspect of a granuloma.

Concetti[10] mentions œdematous infiltration of the lymph spaces and a degeneration of the epithelial cells at various parts of the basal stratum. Here he saw cells with double nucleus and nucleolus resembling the much-discussed coccidial cell inclosures found in carcinoma. The bacteriological examination made by Fede and Concetti did not give noteworthy results. Concetti employed different media and had recourse to animal experiments. Fede reaches the conclusion that the produzione sottolinguale is a purely local lesion of the frenulum linguæ of a uniform character, and does not constitute a peculiar disease. Of particular interest at this point are the two cases of Boccardi, which ended fatally. As a striking clinical illustration, Fede[12] reports the history of an infant, aged ten months, which was in a state of severe malnutrition, etc., at the same time the sublingual tumor was present. The sharp incisors were filed and a mouth-wash of boric acid was used. Under this treatment, lasting four months, the tumor improved greatly, while the general condition of the baby became worse. Ultimately the baby recovered.

In direct opposition to Fede stands Pianese, nephew of U. Cardarelli. His opinion is based on 200 observations, 140 of which were communicated to him by different physicians or by the mothers of the affected babies; 60 cases he observed personally. According to Pianese,[10 11] the form described by Fede differs from that described by Cardarelli, Riga, and himself. His conclusions are that the afta cachettica, or Riga's disease, is an infectious disease which usually occurs in healthy infants of healthy parents toward the third or fourth months of life. The first manifestation is a neoformation on the frenulum linguæ of a pearly-gray color, indolent, of the size of a two-centesimo piece, with an indistinctly defined margin, little elevated over the surrounding tissue. The neoformation may disappear before death, but the baby is not cured. After a certain time the color of the infant changes to a yellowish hue and the baby emaciates. Liver and spleen are enlarged; the bowels are usually constipated. The disease is afebrile and may last for twenty months. If one baby of a family dies from the disease, as a rule the other babies die of it, too. Sometimes one escapes, but the sons of a mother, who as a baby escaped the disease, get it and die. A baby born of an infected mother and nursed by a healthy person escapes, while a baby born of a healthy mother and nursed by an infected person succumbs. The pathological conditions found in three autopsies were bronchopneumonia; fatty degeneration of the liver and of the cortex of the kidney; amyloid degeneration of the spleen. The neoplasm itself is a granuloma showing round-cell infiltration of the derma and deeper in the connective tissue. In some parts cells of an epithelial character are seen. The stratum Malpighii of the neighborhood is slightly hyperplastic. Cultures were taken from the heart, spleen, kidney, lungs, liver, and medulla of the tibia, as well as of the milk of the mother of one of the babies, who came to autopsy. Different media were used. In all the organs, in the milk, and in the sublingual growth the same micro-organisms were found as in the cultures made from them.

In spite of the apparent importance of the disease as described by Pianese, his observations do not seem to have been published *in extenso.* The most surprising feature is that we are left completely in the dark in relation to the nature and characteristics of the micro-organism found. Animal experiments were apparently not made. Some of his statements are directly contradictory to the results of Jannone's experiments. Cannarsa[10] observed the disease frequently partly in the same district as Pianese. Several times he cut the frenulum of healthy children with the same scissors with which he had previously excised the sublingual tumor. The scissors were not sterilized. He never saw an infection and, moreover, nearly all his cases were of a benign nature.

The majority of the authors do not recognize the difference which Pianese tried to establish between the affections described by Riga and Fede. However, there remains more or less difference of opinion as to the nature of the affection, its cause, its prognosis, and as to the conception of its pathological character. In the discussion following the papers[9 10 11] of Fede, Concetti and Pianese, Taura, Dinami, and Tagliaferri stand for the view that the affection is serious, while Nota, Giarrè, and Boccardi, whose cases died, do not believe in the intimate relation between the local lesion and the fatal disease. Lumbardi agrees with Fede, that the sublingual growth may be due to mechanical irritation, and then it is of benign nature. If it comes on spontaneously he regards it, with Pianese, as a manifestation of a general disease, not well known, malignant, and hereditary. The only one of the more recent writers who states that the prognosis is unfavorable in most of the cases is Gazzotti.[13] He reports two cases, one of which died—the surrounding conditions having been very unfavorable—while the other was still living at the time of his publication. None of the other writers, whether they regard the sublingual growth as a purely local affection or put it in some connection with the accompanying general disturbances, is inclined to attribute to it a very serious prognosis.

Concerning the nature of the affection, Gaspardi and Santi[14] reached the conclusion that Riga's malady really is an entity, and, moreover, that it is an infectious disease, in which the coincidence of the lesion under the frenulum and of the general symptoms is not purely accidental. They were led to this opinion by the results which they obtained with the injection of diphtheria antitoxin. The sublingual growth of the infant had resisted the treatment of silver nitrate, the suckling had become more difficult, and the surrounding tissue was slightly infiltrated, while the general health of the baby had suffered considerably. After the first injection of a thousand units, the general condition improved, but locally there was no change. Eight days later the dose was repeated, with good general effects, and now the infiltration around the tumor diminished. Fifteen days later only a little of the tumor was left and the rest disappeared completely after some time. The authors do not regard the affection as a specifically diphtheritic, because the diphtheria antitoxin has proved of value in different infections. In the other two cases they report the complete cure was accomplished by an antiseptic mouth-wash and correction of the diet, the time required being about three months.

Audard[15] recognizes clinically two forms. In the first the sublingual growth is the whole malady; in the second it is associated with serious disease, but it always remains the characteristic symptom. The two cases which he reports (one of Broca and one personal observation) belong to the benign type.

Guida[16] accuses an insufficient function of the mother's breast as the cause of the affection. The frenulum is subjected to considerable friction against the alveolar arch during the prolonged and forcible suckling. Thus a simple abrasion is produced which may lead to ulceration or to chronic hypertrophy of the mucosa of the frenulum. The insufficient nutrition of the baby may cause other disturbances, and so it is easy to understand the different syndromes described as diseases by Cardarelli, Riga, etc.

Gianelli and Memmi[17] and Cappuccio[18] came to the conclusion that the growth is the production of an infectious process; but it will be seen that its influence on the general health is not rated very highly. The mechanism of the infection according to Cappuccio is the following: The stretching forth of the tongue over the lower incisors causes a slight abrasion on the frenulum. In this place, denuded of its epithelium, parasites are deposited, either the usual inhabitants of the mouth or others. Here the parasites may complete their evolution, causing a local reaction, in which the hyperplasia of the connective tissue predominates; the result is a fibroma or papilloma. Or the more virulent parasites penetrate deeper into the tissue, calling forth a more intense reaction, and the result is a granuloma. The sublingual lesion may perhaps occasionally be one of the causes of a general infection; but it is not possible to create the type of a definite infectious disease placing the local lesion and the general infection in relation of cause and effect. The conclusions of Gianelli and Memmi are drawn from a clinical observation of twenty-five cases. In badly fed babies intestinal fermentation and gastro-enteritis set in. Under these conditions the habitual microbes of the buccal cavity may become virulent and start a stomatitis. The stomatitis, which is always of an infectious nature, can bring about a lesion of the sublingual mucosa. While the other lesions disappear without treatment or under the application of an antiseptic mouth-wash, the lesion under the frenulum persists and grows, when to the action of the micro-organisms the mechanical irritation of the lower incisors is added. There is reason to attach much importance to this traumatism; but originally the growth is of an infectious nature. When the bacterial action persists together with mechanical irritation, a granuloma is the result. When the bacterial action is finished, while the mechanical irritation lasts, a papilloma is the product. Of their twenty-five cases, thirteen recovered. They admit that the local lesion can participate in the fatal result in so far as it may interfere with the suckling. But the causes of the atrophy, of the cachexia, of the anæmia, and ultimately of the death, are to be sought elsewhere. So the tumor may disappear, while the baby succumbs to other diseases. The affection does not seem to be contagious. An afflicted baby was fed by a wet-nurse, who gave the breast without precaution to her own baby. This remained perfectly healthy.

The observations of Fedele,[19] Rega,[20] Rossi,[21] Callari,[22] Callari and Phillipson,[23] Olyntho,[24] Israel,[25] Reinbach,[26] Comby,[27] and Brun[28] support the view of Fede and Concetti, that the sublingual affection does not stand in any closer relation to the general disturbances, if, indeed, such are present at all. Some of these authors agree, furthermore, with Fede, that the cause is a mechanical one. Fede, Concetti, Israel, Callari, Callari and Phillipson, Reinbach regard the lesion primarily as a tumor, while the inflammatory changes are secondary. On the other side, Brun, Comby, and Audard regard the tumor as a product of inflammatory processes. Comby describes the lesion under the name of "subglossite diphtheroide," and sees in it a stomatitis limited to the frenulum and of traumatic origin. The degree of traumatism determines whether the result is an ulceration or an hyperplasia of the mucosa.

The interesting data of the descriptions given by Callari and Phillipson, Reinbach and Brun should be reviewed in short. According to Callari and Phillipson, the mass of the tumor consists of connective tissue, which is the direct continuation of the connective tissue of the surrounding mucosa. This mass is not rich in cells. The epithelial layer on the basis of the tumor is thinner than that of the adjoining mucosa, but toward the apex the epithelium becomes decidedly hypertrophic. The papillæ are not marked on the surface. This simple structure is complicated by inflammatory processes. The description of these inflammatory processes coincides as a whole very closely with that given by Fede. The diagnosis is fibroma and not papilloma, because only such neoplasms can be designated papilloma where the formation of the papillæ may be seen macroscopically. The authors incline to the opinion that the tumor represents a congenital deformity. The affection spreads itself in certain districts by means of heredity, and, therefore, remains limited to these districts. To the frequency with which it occurs in southern Italy much importance is attached. In the course of time it can occur in larger family groups, as, for instance, in adjoining districts. In this way a racial tendency can be called into existence, as with other affections. As analogous the authors cite the frequency of the keloid in the Ethiopic races and the occurrence of pigmentation on the nates of many newborns in Japan. In the tumor examined by Reinbach the hyperplastic epithelium sends irregular processes into the underlying tissue. The main part is underneath the epithelium. Here numerous bloodvessels are embedded in stroma of connective tissue rich in cells. The interstitial tissue between the larger vessels, which is rich in cells, too, is in places transformed into thin septa. Thus the type of a cavernous structure is nearly reached. The vessels only sparingly protrude into the papillæ. In the connective tissue circumscribed infiltrations are wanting. Nearly all the cells are connective tissue cells, and only a small fraction is composed of leucocytes. Underneath this layer are fibres of connective tissue, which is not rich in cells of itself, but which shows in several places a typical round-cell infiltration. Between the fibres numerous strings of cells are situated, representing young bloodvessels. Two explanations of the histological findings are possible. One is that the tumor is the result of a chronic inflammatory process; the second and more plausible lets the tumor appear as an angiomatous neoplasm, where the hypertrophy of epithelium is of a secondary nature easily explained by the irritation from the incisors. It is probable that the tumor belongs to the group of the fissural angiomata of congenital origin. The pathological examination in Brun's case was made by Letulle, who found all the strata of the mucous membrane altered. The connective tissue of the mucosa is gorged with bloodvessels, for the most part strongly dilated capillaries. The meshes of the connective tissue are distended by lymph poor in white blood corpuscles. The elongated papillæ are rich in capillaries, which are larger than normal. The epithelial layer is hypertrophic. The centre of the tumor is ulcerated and covered with a pseudomembrane. On the surface a long bacillus was found and in the deeper layers of the pseudomembrane round micrococci, often arranged as diplococci, sometimes in chains; but no micro-organisms were seen in the tissue underneath the pseudomembrane. The growth is a chronic hypertrophy of the mucous membrane of purely inflammatory character. Microbes do not figure in its etiology. The affection which I had occasion to observe resembled macroscopically and microscopically very much the pictures given by Brun.

The microscopic examination furnished the following data: The whole tumor is covered with a thickened layer of pavement epithelium, except in its centre, where the epithelium is absent. The epithelium sends irregular processes of varying length and shape into the underlying tissue. The papillæ ascending between the epithelial processes are composed of connective tissue, which for the most part is not very rich in cells, but well supplied with partly distended capillaries. Occasionally the papillæ are richer in cells and the seat of a round-cell infiltration. The tissue under the epithelium contains a large number of bloodvessels, a part of which are distended. Besides this there are in places a great number of young bloodvessels, represented by strings of cells. The connective tissue is only in rare places comparatively poor in cells, while for the greater part the connective tissue cells are rather numerous. The connective tissue is nearly throughout the seat of a round-cell infiltration of various degrees; besides the round cells occasional polymorphonuclear leucocytes are seen, and in some places nearly all the cells are eosinophiles, which are freely distributed in other places, too. A number of plasma cells complete the picture.*

The centre of the tumor is, as mentioned above, denuded of its epithelium. Here is a mass consisting of detritus, round-cell infiltration, and fibrin. Occasionally a small deposit of fibrin is seen in the tissue adjoining this mass. It remains to be mentioned that all the different cells, mostly round cells, some polynuclear leucocytes, eosinophiles, and plasma cells penetrate into the epithelial layer in different places. In some places they invade the basal strata of the epithelium to a considerable extent, but they rarely wander deeper into the epithelium.

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* According to Pappenheim,[29] the plasma cells are identical with the granulation cells.

The structure of the growth hardly permits of any doubt that it is a product of an inflammatory process. Whether the touching with silver nitrate plays any rôle as to the presence of the large numbers of cells with eosinophile granules is very doubtful, since several days had elapsed between this treatment and the excision; besides the occurrence of eosinophiles, for instance, in nasal polyps is common.[30]

As stated in the beginning, the affection occurs most frequently in southern Italy. Cannarsa, for instance, saw eight or ten cases at the same time among a population of about one thousand inhabitants. Callari and Phillipson, in Rome, found eight cases among six thousand children treated. Gaspardi and Santi state that infection is not so rare in middle and northern Italy as was thought. The affection known under the name of "brufolo" in Egypt, which occurs under the tongue or in the median line of the palate, may belong here. Malinowski[11] thinks he has seen several cases in Poland. From France the cases of Brun, Comby, Audard, and Dufournier[31] are reported; the latter case is somewhat doubtful. From Germany we have the reports of Israel and Reinbach, and from South America that of Olyntho.

The diagnosis does not present any difficulties. None of the cases reported had whooping-cough and only a few coughed slightly if at all.

The etiology of the affection remains obscure. The bacteriological examination thus far has not given satisfactory results. The theory of heredity is not sufficiently proven, neither is the congenital origin. Bad hygienic conditions of the mouth may be of importance. To the mechanical irritation produced by the lower incisors or by the hardening gum an important rôle is attributed. The possibility that any weakened condition of the baby may form a predisposition stands to reason; but some of the infants were apparently in the best state of health. The idea that the sublingual ingrowth is an integral part of a definite serious disease does not seem to be well founded. It seems possible that not all the affections of the frenulum described under the different names of the produzione sottolinguale are of a uniform pathological character, and then the etiology may differ with the different forms.

The age at which the affection occurs seems to be confined to the first two years of life, starting from about the third month, although Olyntho saw it in a four-year-old child. It does not seem necessary to enter upon the different modes of treatment which were proposed. The affection may disappear completely without any particular treatment; but the shortest and simplest way to accomplish a cure seems to be the excision.

The prognosis is favorable.

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PRIMARY RETROPERITONEAL SARCOMA.*

BY HOWARD J. WILLIAMS, A.M., M.D.,
SURGEON TO THE MACON HOSPITAL, MACON, GA.

THE rarity of primary neoplasms in the subperitoneal space and the paucity of the available literature upon the subject induces me to report some cases of retroperitoneal sarcoma. To urge the importance of careful physical examination of all cases presenting persistent digestive disorders, the necessity of early diagnosis in obscure intra-abdominal conditions, and the possible benefits of prompt surgical interference in unusual abdominal growths is my excuse for presenting a paper based upon the records of several fatal cases of these infrequent growths. The diagnosis in two of these cases is based upon exploratory laparotomies; although they were seen too late to be benefited by radical surgery, the histories will be of interest as illustrating

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